Role of Superselective Angiography in the Detection and Endovascular Treatment of Ruptured Occult Arteriovenous Malformations

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Summary

Three cases of occult micro-arteriovenous malformations not identified by cerebral angiography or other imaging modalities were detected by superselective angiography. The first case had a small intracerebral hemorrhage in the superior colliculus, the second had a perimesencephalic subarachnoid hemorrhage, and the third presented with intracerebral hemorrhage combined with massive intraventricular hematoma. While repeated selective cerebral angiography (four-vessel study) was negative, superselective angiography clearly demonstrated each lesion with small early venous filling in accordance with the location of hematoma. Successful superselective embolization with polyvinyl alcohol particles was performed in each micro-arteriovenous malformation by flow-guided microcatheter without postoperative complications. Our experience suggests that superselective angiography is necessary to visualize micro-arteriovenous malformations in patients with cerebral hemorrhage and negative four-vessel angiography. Furthermore, the superselective endovascular approach has the advantage of offering immediate obliteration of the micro-shunt, thereby reducing or eliminating the risk of further hemorrhage.

Introduction

Cerebral angiography has been considered to be the method of choice for the detection of vascular malformations. In 1956, Crawford and Russell[5] introduced the term "cryptic" vascular malformations.

The majority of these cryptic vascular malformations are arteriovenous malformations (AVMs)[1,3,4,9,22,23] that are less than 3 cm in size. Possibly due to their small size, partial or complete thrombosis, obliteration by hematoma, or high vascular resistance of the vascular component, the usual selective four-vessel angiographic investigation is often negative for shunting or early venous filling[7].

The sophisticated superselective angiography with new flexible microcatheters and high resolution digital subtraction angiography (DSA) is one of the techniques to reveal small vascular lesions[2,20,21].

In combination with magnetic resonance imaging (MR) and computed tomography (CT), this procedure also helps to define the origin of hemorrhage[10,18].

In this report, we intend to elucidate the safety and efficacy of superselective angiographic exploration and embolization in patients with occult AVMs.

Case Reports

Case 1

History. A 61-year-old man presented with a sudden onset severe headache, but was conscious and neurologically intact except for mild nuchal dysesthesia. The CT and MR examinations during his first hospital admission showed a 5 mm intracerebral hemorrhage located in the left superior colliculus (figure 1A). Cerebral angiography with DSA on admission was negative, recovery was uneventful, and the patient was discharged with mild nuchal dysesthesia.

Examination. Three months later, the patient was referred to our hospital. On physical examination, findings were normal apart from a minimal nuchal dysesthesia. MR revealed that the left superior colliculus was slightly atrophic but showed no evidence of hemorrhage suggesting absorption of the hematoma (figure 1B). In this case, radiological diagnosis of an intracerebral hemorrhage from cavernoma or micro- AVM was considered.

Operation and Postoperative Course. Under general anesthesia in the DSA suite, left vertebral angiography was first performed, but this

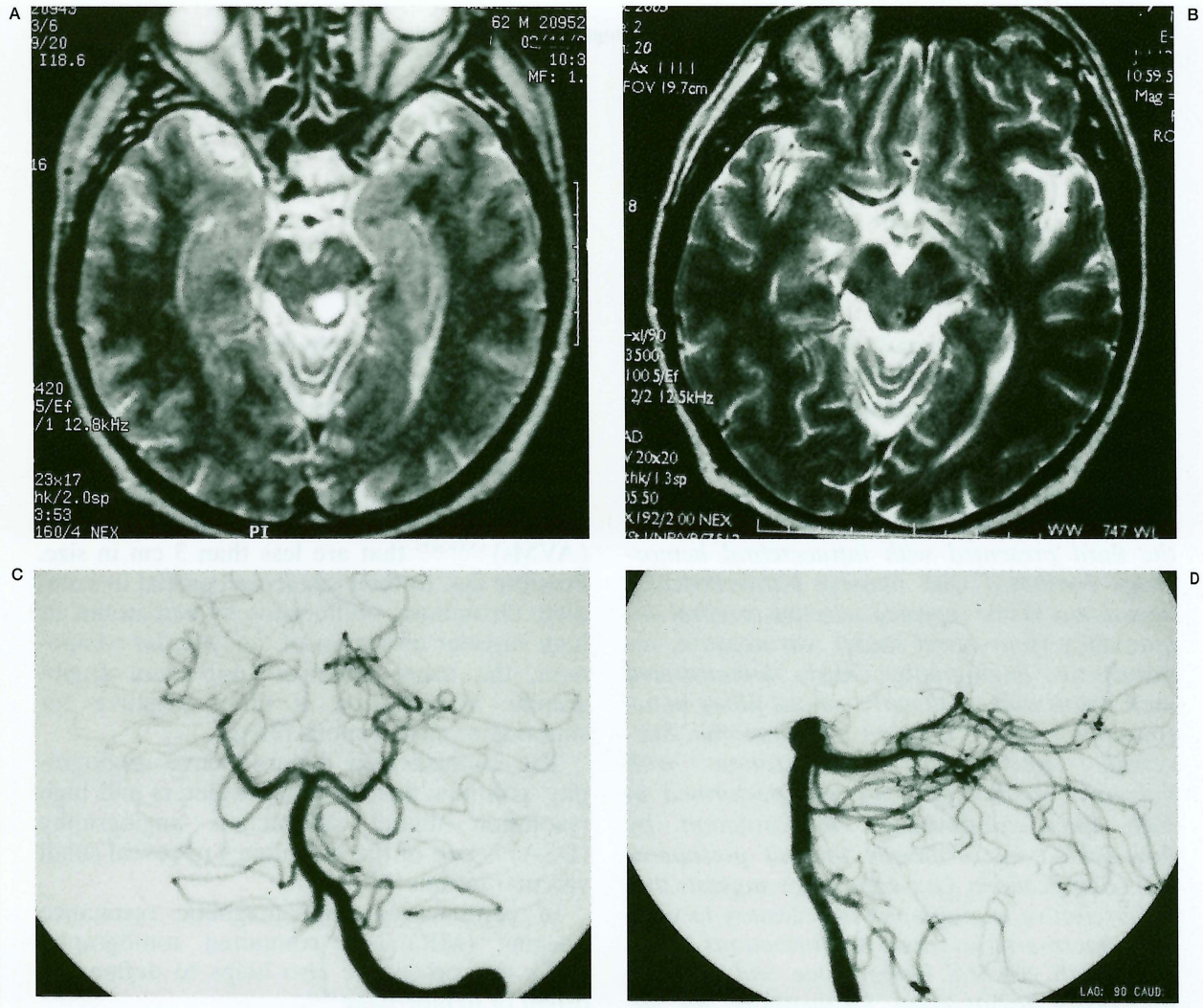


Figure 1 A) Axial T2-weighted MR(TR 3420ms,TE 95ms) study obtained 3 days after onset, demonstrating intraparenchymal hematoma in the superior colliculus of left side. B) Axial T2-weighted MR(TR 3500ms,TE 100ms) obtained 3 months after onset revealed that left colliculus superior was slightly atrophic but no evidence of subsequence hemorrhage suggesting post absorption of the hematoma. C,D) Left vertebral injection (anterior and lateral view) demonstrate no evidence of early venous filling.

did not show any abnormal lesions. (figure 1C,D) Then a No.5.5 French guiding catheter (Valavanis cerebral catheter, Cook,USA) was placed in the distal cervical vertebral artery and superselective catheterization using a No.1.5 French microcatheter (Spinnaker; Boston Scientific, Fremont CA) combined with microguidewire (Transend 0.010-inch; Boston Scientific, Fremont CA) into the distal branch originating from left medial posterior choroidal artery was performed. This superselective angiography demonstrated a small but marked early venous filling draining posteriorly into vein of Galen that was considered a micro-AVM supplied from a single feeding artery (figure 1E,F). Its location corresponded well to the left superior colliculus. Following this microendovascular exploration, a total 1.5mL of contrast material with suspended 45~150µ m polyvinyl alcohol (PVA) particles was injected intermittently. Complete elimination of the early venous filling was then confirmed by superselective angiography with the microcatheter in the same position (figure 1G,H). This patient recovered fully postoperatively and presented some improvement of nuchal dysesthesia. Rebleeding was not observed in follow-up evaluations after 23 months.

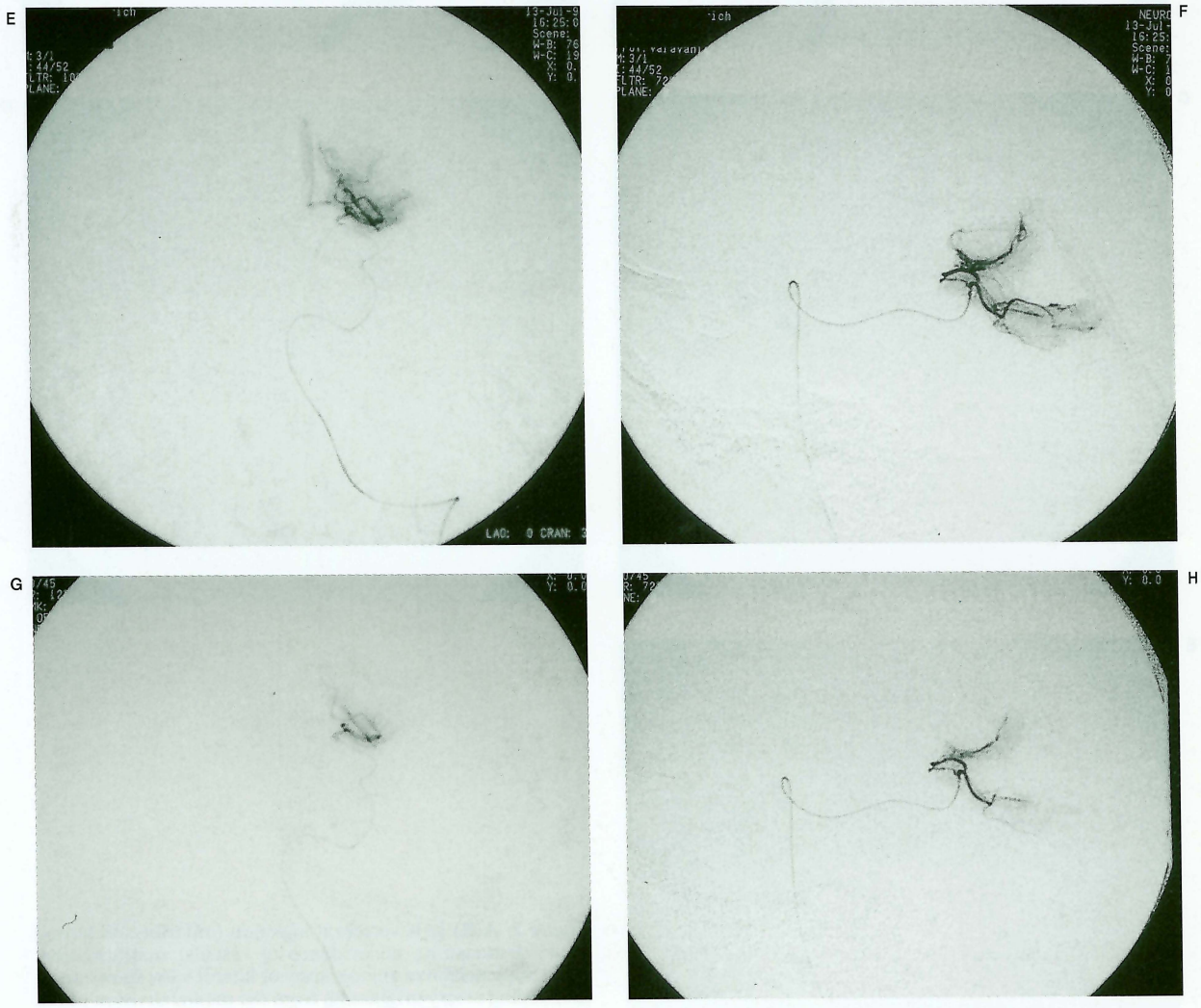


Figure 1 E,F) Superselective angiography (anteroposterior and lateral view) in a parenchymal branch of left medial posterior choroidal artery revealing an early venous filling draining posteriorly into the vein of Galen. G,H) After superselective injection of contrast material with suspended 45~150É m polyvinyl alcohol (PVA), complete elimination of the early venous filling was confirmed by superselective angiography.

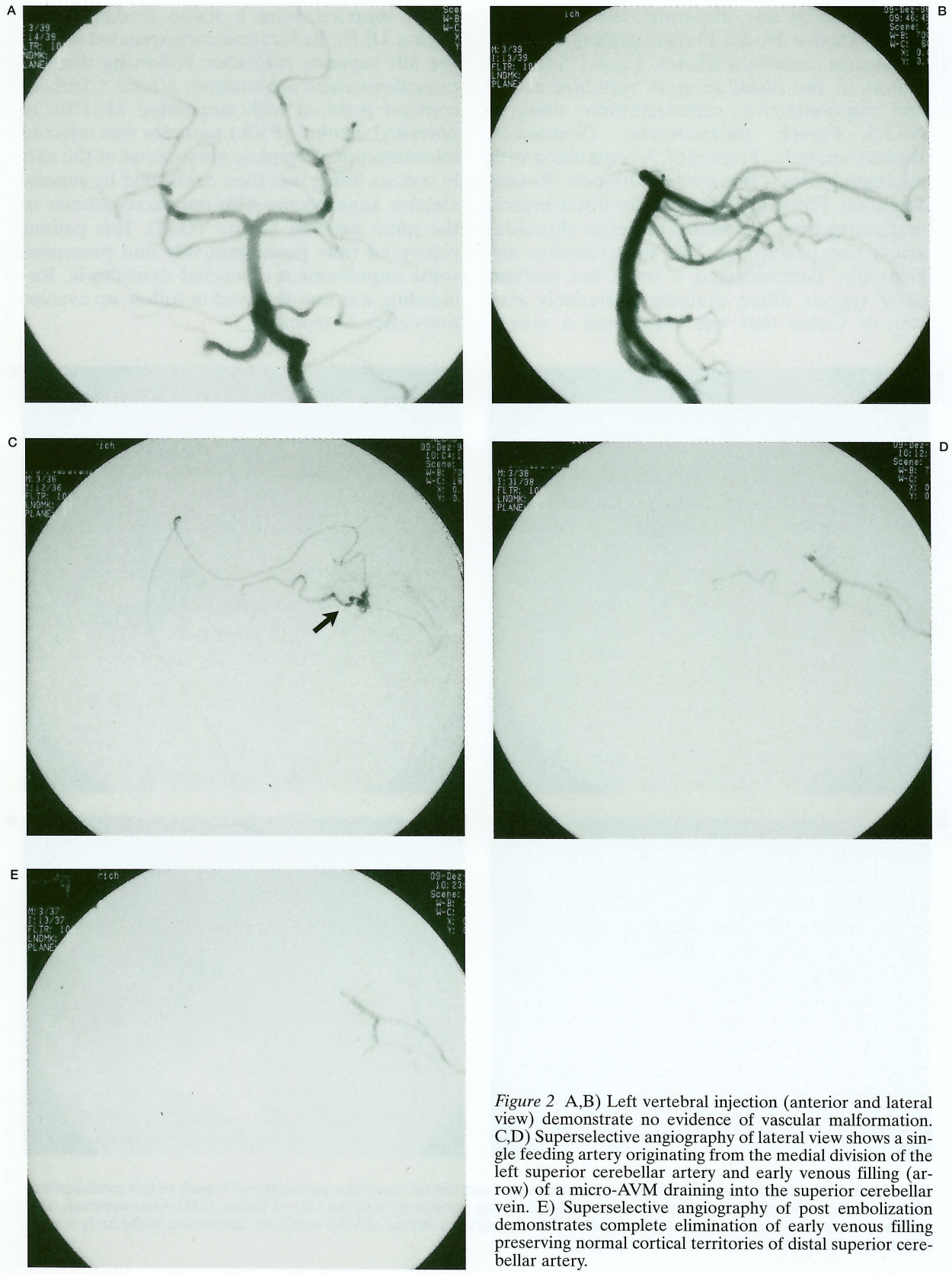


Figure 2 A,B) Left vertebral injection (anterior and lateral view) demonstrate no evidence of vascular malformation. C,D) Superselective angiography of lateral view shows a single feeding artery originating from the medial division of the left superior cerebellar artery and early venous filling (arrow) of a micro-AVM draining into the superior cerebellar vein. E) Superselective angiography of post embolization demonstrates complete elimination of early venous filling preserving normal cortical territories of distal superior cerebellar artery.

Case 2

History. A 32-year-old man presented with severe headache and loss of consciousness. He was transferred to a regional hospital and the CT on admission revealed a subarachnoid hemorrhage located mainly in the periphery of mesencepahlic cistern (Fisher group 2). Cerebral angiography, including vertebral injection, did not show any abnormal vascular lesions. Over the next two days, the patient recovered fully and was neurologically intact.

Examination. Two months later this patient was transferred to our hospital in order to explore the source of the subarachnoid hemorrhage. On admission, the patient was neurologically intact. An axial T2-weighted image of MR at the level of the right mesencephalon showed only a heterogeneous hyperintense lesion suggesting the existence of hemosiderin and methemoglobin.

Operation and Postoperative Course. Under general anesthesia in the DSA suite, the patient first underwent cerebral angiography and this selective angiography showed no abnormal vascular lesion (figure 2A,B). A subsequent superselective angiography using a 1.5 French microcatheter (Spinnaker; Boston Scientific, Fremont CA) into a small feeding artery originating from the medial division of the left superior cerebellar artery finally revealed a micro-AVM draining into the superior cerebellar vein and precentral cerebellar vein (figure 2C,D).

After confirmation of this early venous filling with superselective angiography, a total of 1.0 ml of contrast material with suspended 45~150μ m polyvinyl alcohol (PVA) particles was intermittently injected and early venous filling was eliminated. (figure 2E). The patient recovered fully without any neurological deficit and postoperative MRI examination revealed no evidence of abnormal lesions except for the deposition of hemosiderin in the mesencephalon. No rebleeding was observed in follow-up evaluation at 30 months.

Case 3

History. A 15-year-old male presented with a first epileptic attack and loss of consciousness. He was transferred to a regional hospital and the CT on admission revealed a massive intracerebral hemorrhage at the trigonal part of left lateral ventricle. (figure 3A,B) Cerebral angiography did not show remarkable abnormal vascular lesions. Over the next four days, the patient recovered and was neurologically intact except for partial visual field defect.

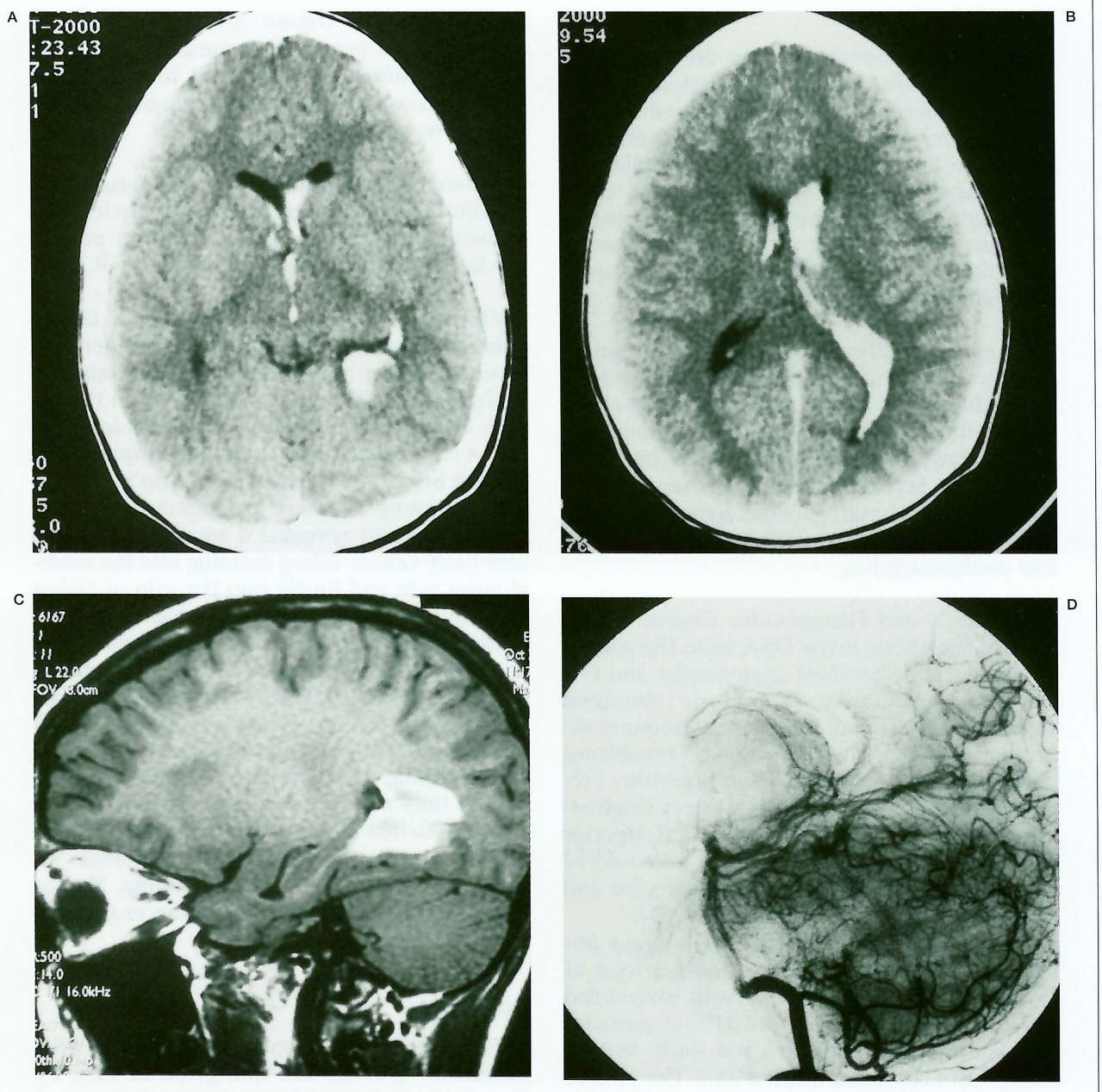
Examination. Six days later this patient was transferred to our hospital. MRI disclosed a hematoma located mainly at posterior part of the left parahippocampal gyrus (figure 3C).

Operation and Postoperative Course. Under general anesthesia, the patient first underwent cerebral angiography and this selective angiography did not show any vascular lesion (figure 3D). However, superselective angiography using a 1.5 French microcatheter (Spinnaker; Boston Scientific, Fremont CA) into a posterior hippocampal branch originating from the left P3 segment revealed a small but remarkable early venous filling draining into the medial atrial vein and finally into the vein of Galen confirming the presence of a micro-arteriovenous fistula composed of a single feeder and single draining vein (figure 3E,F). Subsequently, a total 1.4 ml of contrast material with suspended 45~150μ m polyvinyl alcohol (PVA) particles was injected intermittently and early venous filling was eliminated (figure 3G). The patient fully recovered without any newly neurological deficit and postoperative MRI examination revealed no evidence of abnormal lesions except for the initial hematoma. No rebleeding was observed in follow-up evaluation at 11 months.

Discussion

The designation "cryptic arteriovenous malformation" (AVM) was first given in 1956 by Crawford and Russel 5 to a cluster of abnormal arteriovenous connections measuring less than 2 to 3 cm maximum. In 1980, Schlahter et al [16] reported a case that presented a right parietal hematoma and a left tentorial AVM. Although no vascular lesion was seen around the hematoma on an angiography, the existence of a small AVM was proven pathologically in the evacuated hematoma.

Arteriovenous malformation is usually defined by early venous filling on conventional angiography. However, if the malformation presents with hemorrhage, its precise vascular

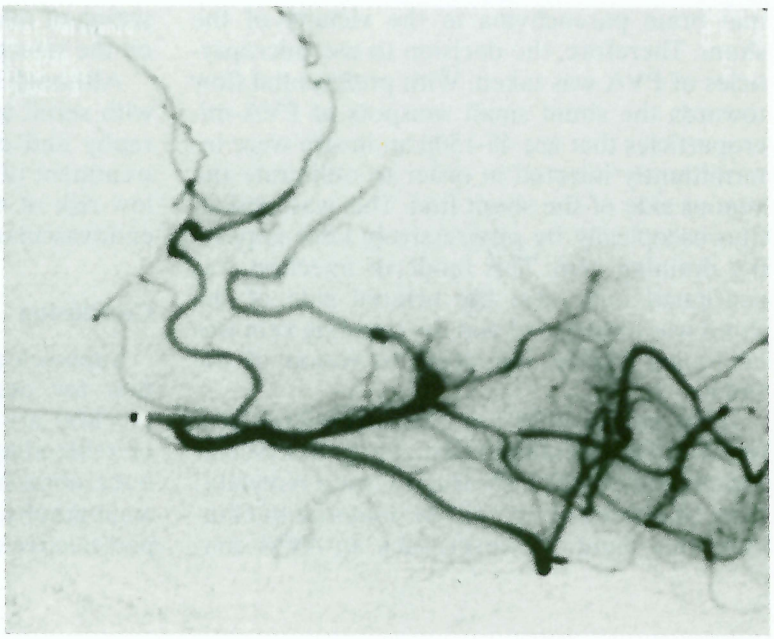


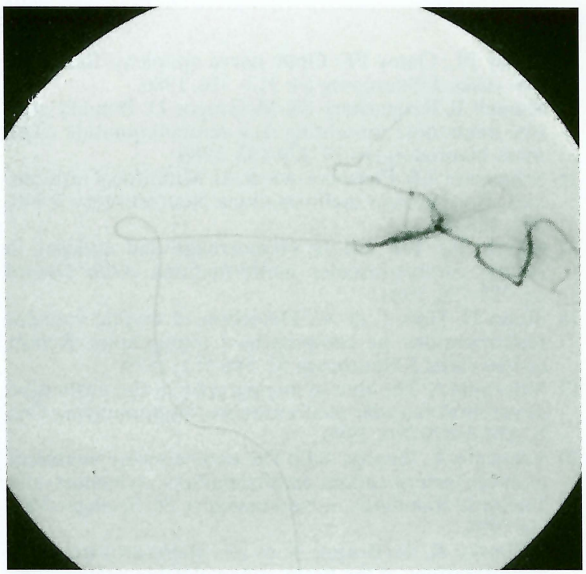
structure is compressed by the hematoma and intranidal vascular resistance increases. Therefore, its venous drainage might be delayed and this type of small AVM may become a so-called angiographically occult AVM that means anatomically present but angiographically invisible. These occult vascular malformations are not the benign entity they were previously thought to be because they can cause recurrent hemorrhages and persistent neurological deficits [7,8,11,12,13,14,17,22]. Additionally, some of the cases that present with subarachnoid hemorrhage (SAH) of unknown origin, frequently observed as a perimesencephalic SAH, might be also categorized to this group of angiographically occult AVM.

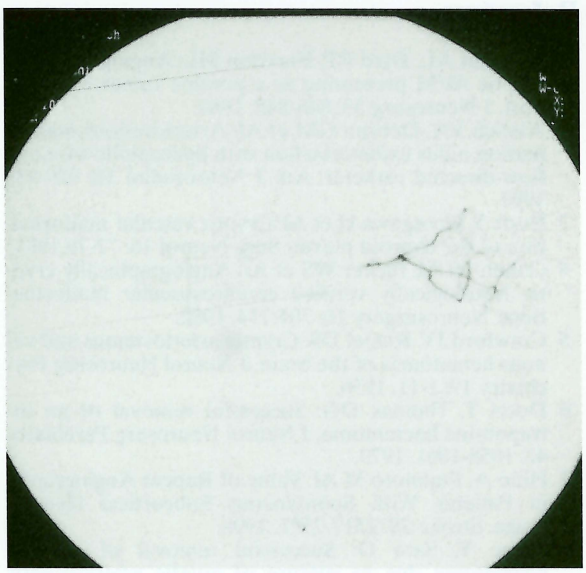
Modern sophisticated microcatheter techniques under high resolution digital subtraction angiography can provide the ability to diagnose micro-AVMs that can be embolized with particle PVA [2,19,20,21].

The decision to use microparticles of PVA

Figure 3 A,B) CT on admission revealed a massive intracerebral hemorrhage at the trigonal part of left lateral ventricle. C) MRI showed that hematoma located mainly at posterior part of the left parahippocampal gyrus. D) Vertebral angiography late arterial phase does not reveal any abnormal vessel. E,F) Angiography through the microcatheter demonstrates early opacification of the medial atrial vein with drainage to the vein of Galen. F) The microcatheter tip is located just proximal to the origin of the posterior hippocampal branch. G) Superselective angiography after PVA injection shows complete elimination of early venous filling with preserving of distal normal cortical supply.







instead of NBCA (N-Butyl Cyano Acrylate) in all three cases was based on following observations;

1. The tip of the microcatheter was positioned within the terminal segment of the feeding artery but still outside the nidus. The microcatheter could not be advanced further due to the small diameter of the feeding artery. On superselective angiography parenchymal stain was observed but appeared later than the opacification of the shunt. Therefore, there was preferential flow towards the shunt.

2. The flow through the shunt was relatively slow with rather prolonged visualization of the small early draining vein.

3. No arterial or venous pseudo-aneurysm was present in the nidus, which would represent a contraindication to the use of microparticles.

Use of even highly diluted NBCA would result in occlusion of the terminal segment of the feeding artery, which was also supplying nor-

mal brain parenchyma in the vicinity of the shunt. Therefore, the decision to use microparticles of PVA was taken. With preferential flow towards the shunt small sunspots of PVA-microparticles that are 45-150μ m in size were intermittently injected in order to obliterate the venous side of the shunt first. This was evident fluoroscopically by progressively later appearing draining vein. This mode of injection was continued until also the arterial side of the shunt was obliterated and the draining vein was no longer visible, indicating occlusion of the shunt.

If the micro-AVM has a relatively high flow shunt and low vascular resistance of its vascular component , NBCA (N-Butyl Cyano Acrylate) must be chosen as an embolic material to eliminate this shunt. However, such an AVM consisting of relatively high flow shunt can be seen on the selective angiography.

Although it is necessary to follow the patient with serial angiography, superselective angiography and embolization allow the immediate treatment of malformations with an acceptably low risk of morbidity and mortality related to endovascular procedure.

Conclusion

Superselective angiography and embolization for occult arteriovenous malformations (AVMs) are safe and effective for prevention of rebleeding. In patients presenting with intracerebral bleeding and negative conventional angiography, further investigation including superselective angiography is recommended.

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EDITORIAL COMMENT

This is an interesting and controversial paper. The authors present three patients who had intracranial hemorrhage and negative cerebral angiograms. The first patient had a small hematoma in the brain stem, another had an intracerebral hematoma near the trigone of the lateral ventricle and one had a perimesencephalic subarachnoid hemorrhage. Superselective angiography under general anesthesia revealed "slow flow" arteriovenous shunt at three months in one patient, two months in the second patient and 10 days in the third patient. These arteriovenous shunts were not evident on the cerebral angiograms despite using general anesthesia. These micro-arteriovenous fistulas were embolized with particles and the immediate results showed that the shunts were eliminated.

The controversy of the paper relates to the natural history of these arteriovenous shunts evident only on superselective angiography. The authors have designated these to be "slow flow shunts". The natural history of these "slow flow shunts" may be different from the natural history of brain arteriovenous malformations. At present, there is no natural history data on arteriovenous shunts that are only evident on superselective angiography. These shunts may have developed as a result of ischemia or infarction related to the initial bleed and may not have the bleeding risk that is associated with micro-arteriovenous malformations evident on cerebral angiography [1].

The management of these unique cases is also open to debate. Recanalization is known to occur after the use of PVA particles in the embolization of cerebral arteriovenous malformations [2]. The rationale for the use of particles in these cases is clear since a normal parenchymogram was evident on the superselective study. The authors have not provided follow-up angiography that would need to include repeat superselective studies.

The authors have recommended that patients presenting with intracerebral bleeding and negative angiography should go on to superselective angiography. In patients with unexplained intracerebral hemorrhages, most centers do delayed angiography after the hematoma has resolved [3]. Early and delayed MR complements this protocol by detecting underlying mass lesions and cavernomas. Until there is more evidence to support the authors' unique approach, our group will continue to use high quality cerebral angiography to investigate unexplained intracerebral hematomas and use superselective angiography in selected cases where any early drain vein is evident [4].

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